Annals of Clinical and Analytical Medicine
Case Report

Integrated treatment for camptocormia, parkinson's disease and bruxism: A case report

Elisa Calisgan[1], Betül Akyol[2], Cristina Frange[3]
[1] Department of Physiotherapy and Rehabilitation, Faculty of Health Science, Kahramanmaras Sutcu Imam University, Kahramanmaras, Turkiye
[2] Department of Physical Education and Sport, Faculty of Sport Science, Inonu University, Malatya, Turkiye
[3] Department of Neurology and Neurosurgery, Federal University of Sao Paulo (UNIFESP); Sleep Physio, São Paulo, Brazil

Abstract

One of the most common symptoms of Parkinson's disease (PD) is abnormal posture. Camptocormia, which is rarely observed in patients with PD described the extreme forward-bending trunk position known as "bent spine syndrome", a condition in which the trunk flexes abnormally when standing or walking but disappears in the supine position. This clinical case includes a 73 year-old male patient diagnosed with PD and sleep bruxism bruxism, presenting Camptocormia, symptoms of tremor, back pain, rigidity, bradykinesia, postural instability, limited mouth opening, decreased facial expression, and fatigue. Width of the mouth opening, mobility, functionality, range of motion, fatigue severity, language, speech and swallowing functions, and quality of life were evaluated before and after treatment. In this study, a detailed evaluation of the patient and a physiotherapeutic treatment protocol were created and performed along with pharmacological treatment focused on diminishing pain, improving mouth opening, swallowing functions and the tone of facial muscles, decreasing tone of abdominal muscles and stretching abdominal fascia, strengthening of the back extensor muscles and core muscles, and improving balance.

Keywords

Camptocormia, Physiotherapy, Rehabilitation, Parkinson Disease, Bruxism

DOI: 10.4328/ACAM.22384 Received: 2024-08-23 Accepted: 2024-09-30 Published Online: 2024-10-19 Printed: 2024-10-20 Ann Clin Anal Med 2024;15(Suppl 2):S102-106
Corresponding Author: Elisa Calisgan, Department of Physiotherapy and Rehabilitation, Faculty of Health Science, Kahramanmaras Sutcu Imam University, Kahramanmaras, Turkiye.
E-mail: elisacalisgan@ksu.edu.tr P: +90 534 246 24 71
Corresponding Author ORCID ID: https://orcid.org/0000-0003-4710-9540
Other Authors ORCID ID: Betül Akyol, https://orcid.org/0000-0002-3836-1317 · Cristina Frange, https://orcid.org/0000-0002-5435-3579

Introduction

Parkinson's disease (PD) is a neurodegenerative disorder that primarily impacts dopaminergic neurons in the substantia nigra, leading to a reduction of dopamine in the basal ganglia-thalamocortical circuits. As a result, both motor (i.e., tremor, back pain, rigidity, bradykinesia, postural instability, limited mouth opening, decreased facial expression), and non-motor symptoms such as fatigue and sleep disorders (i.e., sleep bruxism) are observed. PD can also manifest with abnormal postures like Camptocormia, neck dystonia, and dropped head syndrome. The evaluation criteria for Camptocormia are brain CT (Computed Tomography), and MRI (Magnetic Resonance Imaging), blood chemistry and neurological evaluation [1].

Camptocormia, also referred to as a bent spine syndrome, is a major complication of advanced PD. It is known as an abnormal/non-fixed flexion of the thoracolumbar spine, observed on standing or walking, with remission on supine position. The International Parkinson and Movement Disorders Society Study Group defined Camptocormia as a neromuscular disorder, a forward bending angle of >30° [2]. Lower Camptocormia (L1-Sacrum, hip flexion) can be defined as a bending angle of ≥30°, and upper Camptocormia (C7 to T12-L1) as a bending angle of ≥45° [3]. The pathogenesis of Camptocormia is still unknown, but the proposed hypothesis relies on a centrally caused abnormality in systems regulating trunk posture [4], and a primary idiopathic axial myopathy related to trunk dystonia [5]. This imbalance of posture due to the weakness of paraspinal and extensor muscles (massive fatty infiltration of paravertebral muscles) and overactivity of abdominal muscles generates a trunk forward flexion posture, bent knees, flexed elbow and ankle, with increasing risk of falls. Camptocormia designates a forced posture with a forward-bent trunk, which triggers extending the neck which may cause weakness of the extensor muscles of the neck, temporomandibular joint dysfunction and sleep bruxism. Sleep bruxism is a repetitive jaw-muscle activity characterised by clenching or grinding of the teeth and/or by bracing or thrusting of the mandible. In turn, sleep bruxism may change craniocervical posture increasing neck angle [2, 6].

There is no pharmacological treatment for Camptocormia, but frequently used are antiparkinsonian drugs (e.g., levodopa, dopamine agonists, anticholinergics and amantadine) which partly covers the treatment. Non-pharmacological treatment are deep brain stimulation (subtathalamic nuclei), corsets for improving kyphosis, and physiotherapy. The patients, who used deep brain stimulation into subthalamic nucleus, underwent clinical assessment utilizing a continuous waveform width of 60 µs in monopolar stimulation, along with amplitude increments of 0.2-0.5 V at a frequency of 130 Hz [1, 5].

This case report presents a combined physiotherapy management for a patient with concomitant PD, Camptocormia and Sleep bruxism.

Case Report

Patient Information

A male patient, 73 years-old, with Camptocormia (thoracolumbal bending, Figure 1), Hoehn and Yahr Scale range: 1.5-2.5, indicating bilateral involvement without impairment of balance, axial involvement and mild bilateral disease with recovery on pull test, attending the Ataturk University Hospital was invited to participate in the study. Ethical approval was not required. Written patient consent to publication of this case history was obtained.

The patient had comorbid bruxism and presented with spasms in both masseter, orbicularis and trapezius muscles, with complaints of neck and back pain and difficulty opening the

Table 1. The quality of life, functionality, fatigue and swallowing functions results before-after of rehabilitation

Evaluation scales	Pre-Rehab	Post-Rehab	Mean Difference
PDQ-39 (point)	92.08	61.09	30.99
Width of the mouth opening (cm)	5.1 cm	5.8 cm	0.7 cm
Fatigue Severity Scale (FSS) (point)	7.00	4.55	2.45
Jaw Restriction Scale (point)	53.00	31.00	22.00
SWAL-QOL (point)	48.00	178.00	130.00

Table 2. Upper and Lower extremity range of motion (ROM) measurement results before-after of rehabilitation

Joints of the body	Movement	Pre-Rehab Right	Pre-Rehab Left	Post-Rehab Right	Post-Rehab Left
Shoulder	Flexion	70°	70°	120°	120°
	Extension	30°	30°	45°	45°
	Abduction	60°	60°	110°	110°
	Internal rotation	70°	70°F	70°	70°
	External rotation	60°	60°	80°	80°
Elbow	Flexion	90°	90°	120°	120°
Wrist	Flexion	60°	90°	90°	90°
	Extension	20°	20°	60°	60°
Finger	Flexion	30°	30°	45°	45°
Hip	Extention	20°	15.2°	49.1°	46.3°
Knee	Extention	20.3°	20.1°	42.9°	42.8°

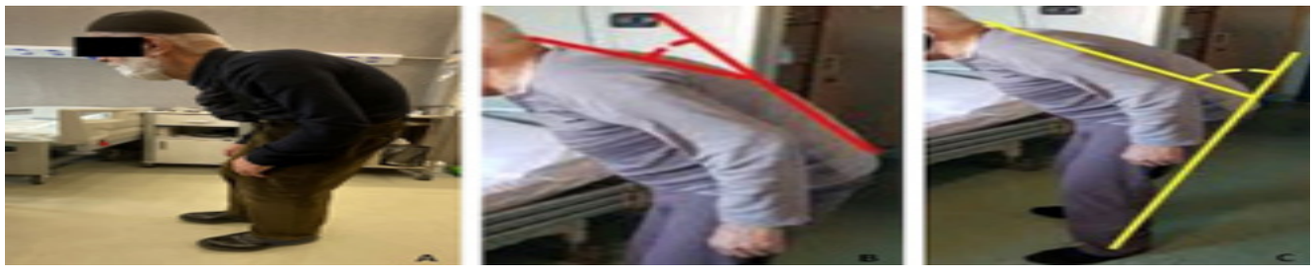

Figure 1. A. Lateral view of the patient; B. Illustration of lower thoracic flexion angle of 45.9º; C. Illustration of upper lumbarvertebrae flexion angle of 47.3º

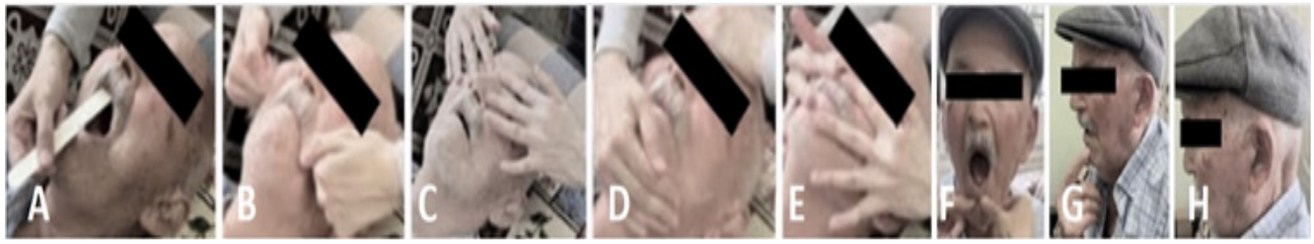

Figure 2. A., B., C., D., E., Hands-on approach on facial and neck muscles; F., G., H. Rocabado exercises

Figure 3. Lateral view of the patient after treatment

mouth due to pain.

Clinical Findings

In laboratory examination, hemogram, erythrocyte sedimentation rate (ESH), liver and kidney function tests, alkaline phosphatase, creatine kinase (CK), calcium, phosphorus, osteocalcin, C-reactive protein (CRP), rheumatoid factor (RF), anti-nuclear antibody (ANA), thyroid function tests (TFT) and parathyroid hormone (PTH) levels were normal. The patient's Vitamin D (25-OH D3) level was 7.4ng/mL (20-80ng/mL). An endocrinologist was consulted for possible etiological reasons of hipodeficency and vitamin D replacement started. The patient sought consultation with psychiatry to explore potential causes like conversion disorder, exhibited no signs of psychopathology. Paravertebral muscle electromyography (EMG) was not performed for axial muscle myopathy. There was no clinical sign of myopathy, as diagnosed by the physician, and the resultof MRI was mild radicular involvement at L5-S1 level. Measurement of spine angles were performed through goniometer, and the flexion angle of lower thoracis before treatment was 45.9°, while the flexion angle of upper lumbarvertebrae was 47.3º (Figure 1). Angle of the mouth opening was measured through a mandibular goniometer. Measurement of the mouth opening was performed by the distance of the edges of the frontal incisors using a ruler, and was 5.1 cm at baseline.

Diagnostic Assessment

Evaluation scales were

i) Parkinson Disease Questionnaire (PDQ-39), which assesses how often people with Parkinson's experience difficulties across 8 dimensions of daily living. PDQ-39 consists of 39 multiple-choice questions, covering patients' mobility, daily living activities, emotional well-being, societal stigma, social support, cognitive function, communication, and physical impairment. Each dimension's total score ranges from 0 (indicating no difficulty) to 100 (representing constant trouble). Lower scores are indicative of better health, while higher scores suggest a decline in the quality of life [1];

ii) Fatigue Severity Scale (FSS), a 9-item questionnaire designed to assess fatigue. Each item is rated on a scale from 0 to 7. To determine the overall fatigue level, the total score is divided by 9. A mean score <4 indicates no fatigue, while a mean score ≥4 suggests excessive fatigue [2];

iii) Jaw Restriction Scale (JRS) assess the limitation of the jaw movement, which is very common symptom of bruxism. It consists of items that evaluate everyday mouth function, such as chewing, yawning, swallowing, and smiling. The score is from 0 to 10 for each item, where 0 represents no restrictions, and 10 indicates severe limitations. The overall score ranges from 0 to 80. Score of 0 reflects no jaw restrictions and excellent condition, and a score of 80 signifies complete restriction of jaw movement [2];

iv) Swallow Quality of Life Questionnaire (SWAL-QoL) were used for evaluation of language, speech and swallowing functions, quality of life, and superficial postural situation. The SWAL-QoL measures the impact of a swallowing disorder on a patient's quality of life, gathering information directly from the patient. It comprises 44 questions that assess various aspects of quality of life. Each question is assigned a score ranging from 1 to 5, with 1 representing "completely correct" and 5 indicating "not at all correct." As the score increases, it indicates an improvement in swallowing functionality [3].

Therapeutic Intervention

A patient with a nine-year record of PD, who had previously exhibited a positive reaction to levodopa, came for assessment due to progressively incapacitating motor fluctuations and

notable camptocormia. This patient underwent pharmacological, which remained unchanged dose after rehabilitation, and non-pharmacologial treatment. Pharmacological included liquid and oral L-DOPA/dopadecarboxylase inhibitor, at dosage of 200/50 mg regularly.

Physiotherapy and rehabilitation happened 4 times a week, during 8 weeks (pre-treatment in one session and post-treatment evaluation in one session), comprising 32 sessions : i) 30 sessions of transcutaneous electrical nerve stimulation please correct: here vertebral region to pain relief. Compex Chattanooga® TENS device was used. A single large passive electrode (258 cm2) and two active electrodes (25 cm2 each) were chosen, totalling 12 electrodes. During the procedure, the passive electrode was positioned on the thoracic region, while the active electrodes were placed on the lumbar paravertebral and multifidus motor points. The device settings consisted of 5 seconds of stimulation followed by 5 seconds of no stimulation, with a frequency of 50 beats per second. The voltage was gradually increased until achieving a level of submaximal contraction. This application protocol was sustained for a duration of 15 minutes; ii) 30 sessions of hot packs on back; iii) 15 sessions of therapeutic ultrasound, frequency 3 MHz, a duration of 5 minutes, on bilateral transversus abdominis and multifidus muscles; iv) 30 sessions of rehabilitation exercises consisting of waist-neck exercises, strengthening of the back extensors, stretching the abdominal fascia, stretching the pectoralis muscles, strengthening the core muscles, balance and walking exercise; v) hands on approach on facial and neck muscles, such as Proprioceptive neuromuscular facilitation exercises (PNF), myofascial release techniques (Figure 2). One hour session was composed of TENS for 15 mins, ultrasound for 5 mins, PNF and myofascial release for 15 mins and 3 of the Rocabado exercises (Figure 2), rehabilitation exercises for 20 mins and hot pack for 5 mins at the end.

He was prescribed a spinal orthosis, Jewett type hyperextension brace. Also, an inelastic 8-shaped bandage applied to the back, with the midpoint of the dorsal apex of the kyphosis. As he could not straighten his back, a rope was passed through the middle point of the bandage, and tied to the waist belt at two points. Thanks to this pull, he perceived to have an upright posture (Figure 3). Elastic exercise bands take the place of elastic rope over time. While the most resistant band is used at the beginning, it is later switched to the band that gives less resistance. The patient was instructed to use the orthosis throughout the day, with the exception of the exercise period and overnight sleep. For the exercise routine, he was directed to wear the 8-shaped corset while maintaining an upright posture.

Follow-up and Outcomes

At baseline, PDQ-39 was 92.08; FSS was 7.00, JRS e was 53; and the score of SWAL-QoL was 48. After treatment, the width of the mouth opening improved from 5.1 cm to 5.8 cm. Measurement of the difficulties in daily living (PDQ-39) decreased from 92.08 to 61.09, indicating improvement of quality of life; limitation of jaw moment improved from 53 to 31, based on JRS. Taken together these observations indicated improved functionality, reduced fatigue, better swallowing functions and enhanced quality of life, after treatment (Table 1).

Following the intervention, significant improvements were observed in the range of motion

for both the upper and lower extremities via goniometer. This enhancement contributes to an improved patient posture and functionality, thereby increasing overall capacity (Table 2).

The bending angle of lower thoracic region decreased from 45.9º to 25.4º, while the bending

angle of the upper lumbar vertebrae improved from 47.3º to 5.9º (Figure 3).

Discussion

This case report presented a physiotherapeutic and rehabilitation program of treatment, aligned with pharmacotherapy, of a patient presenting with Camptocormia, PD and Sleep bruxism. Schroeteler et al. (4) presented 3 cases in which a high frame walker with forearm support was employed to alleviate back pain and enhance walking distance. Conversely, De Seze et al. (5) applied thoraco-pelvic anterior distraction orthosis and physiotherapy to 15 in patients with Camptocormia, resulting in increased lumbar lordosis, reduced pain, and improved quality of life. Additionally, Ye et al. (6) reported a single case demonstrating the resolution of ambulatory disability and flexion posture on a permanent basis through the use of a cruciform anterior spinal hyperflexion device and back extension strengthening exercises.

Although some camptocormia treatment studies have been presented as case reports, Capecci et al. (7) conducted a single-blind, randomized controlled trial involving 20 patients with Parkinsonism who experienced postural deformities. In their study, 7 patients received tailored postural rehabilitation, and 6 patients received a combination of tailored postural rehabilitation and Kinesiotaping. Similar to our study, they also observed improvements in trunk posture, both in sagittal and coronal angles. However, our study differs from Capecci et al. (7) that was focused on tactile stimulation and postural re-education, while we emphasized stretching of facial muscles and back extensor strengthening exercises. In 2005, Sinaki et al. published a study wherein individuals with spinal osteoporosis were provided with a weighted kypho-orthosis accompanied by exercise. The results demonstrated enhancements in kyphotic posture, body balance, gait velocity, and cadence. It's important to note that the underlying causes of Camptocormia in spinal osteoporosis and Parkinsonism are different significantly. Their study were focused on individuals suffering from Camptocormia associated with osteoporosis (8). The results similar to those of our study showed enhancements in kyphotic posture, body balance and functionality.

Conclusion

This case report presented a physiotherapeutic approach to Camptocormia, PD and sleep bruxism. The physiotherapy and rehabilitation protocol used demonstrated the effectiveness of therapeutic exercise in enhancing activities of daily living and alleviating motor symptoms associated with PD and Camptocormia and bruxism. As a result, we propose that

conservative approaches, like hands-on approach, backpack-wearing or back extensor strengthening exercises should be considered as the initial course of action, conservative treatment, rather than invasive methods such as orthopedic surgery. With the enhancement of posture, the patient improved forward head position, experienced a decrease in muscle imbalance leading to TMJ dysfuncion and probably bruxism, as well as the masseter, hyoid and pterygoid muscles, attributable to the adjustment of the chin posture. We believe that this case report will open a venue in treatment possibilities, being useful in helping clarifying the issue of PD, Camptocormia and Sleep bruxism, alleviating the symptoms for a better quality of life.

Scientific Responsibility Statement
The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement
All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or compareable ethical standards.

Conflict of interest
The authors declare that there is no conflict of interest.

References
1. Azher SN, Jankovic J. Camptocormia: Pathogenesis, classification, and response to therapy. Neurology. 2005;65(3):355-359.
2. Ohrbach R. Diagnostic Criteria for Temporamandibular Disorders: Assessment Instruments. 2016;15.
3. Finizia C, Rudberg I, Bergqvist H, Rydén A. A cross-sectional validation study of the Swedish version of SWAL-QOL. Dysphagia. 2012;27(3):325-35.
4. Schroeteler FE, Fietzek UM, Ziegler K, Ceballos-Baumann AO. Upright posture in parkinsonian camptocormia using a high-frame walker with forearm support. Mov Disord. 2011;26(8):1560-1561.
5. de Seze MP, Creuze A, de Seze M, Mazaux JM. An orthosis and physiotherapy programme for camptocormia: A prospective case study. J Rehabil Med. 2008;40(9):761-765.
6. Ye BK, Kim HS, Kim, YW. Correction of camptocormia using a cruciform anterior spinal hyperextension brace and back extensor strengthening exercise in a patient with Parkinson disease. Ann Rehabil Med. 2015,39(1):128-132.
7. Capecci M, Serpicelli C, Fiorentini L, Censi G, Ferretti M, Orni C, et al. Postural rehabilitation and Kinesio taping for axial postural disorders in Parkinson's disease. Arch Phys Med Rehabil. 2014;95(6):1067-1075.
8. Sinaki M, Brey RH, Hughes CA, Larson DR, Kaufman KR. Significant reduction in risk of falls and back pain in osteoporotic-kyphotic women through a Spinal Proprioceptive Extension Exercise Dynamic (SPEED)program. Mayo Clin Proc. 2005;80(7):849-855.

How to cite this article:
Elisa Calisgan, Betül Akyol, Cristina Frange. *Integrated treatment for camptocormia, parkinson's disease and bruxism: A case report. Ann Clin Anal Med 2024;15(Suppl 2):S102-106*